Pseudoaneurysm Formation Following Intrasphenoid Rupture of an Idiopathic Intracavernous Carotid Artery Aneurysm: Coil Migration and Early Recurrence after Endovascular Treatment

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Summary

Intrasphenoid rupture of a non-traumatic aneurysm of the cavernous carotid artery is rare. We describe a patient in whom this condition manifested with epistaxis and led to the formation of a pseudoaneurysm occupying the right sphenoid sinus. The lesion recurred after repeated attempts at conservative endovascular therapy. Eventually the patient was treated with endovascular occlusion of the right internal carotid artery. Our report emphasizes the relapsing behaviour of a non-traumatic aneurysm of the cavernous portion of the internal carotid artery ruptured into the sphenoid sinus.

Introduction

Aneurysms of the cavernous portion of the internal carotid artery account for approximately 2-9% of all intracranial aneurysms, and can be classified as idiopathic, infectious and traumatic [1]. Idiopathic lesions are true aneurysms and are contained by layers of normal arterial wall. By contrast, traumatic and infectious lesions are pseudoaneurysms. The pathogenesis of pseudoaneurysms includes partial vessel injury and formation of an hematoma, whose unclotted portion is filled by circulating blood in continuity with the arterial lumen and is surrounded by a fibrous capsular wall. True aneurysms in this site have a low risk of rupture, and are asymptomatic in up to 34% of cases [2]. The most common symptoms are compressive (including the classical cavernous sinus syndrome, headache, retro-orbital or facial pain), whereas vascular symptoms (among which rupture, ischemic phenomena, formation of carotid cavernous fistulas) are rare. Subarachnoid hemorrhage occurs with a very low rate, approximately 0.4% per patient-year [3]. Pseudoaneurysms of the intracavernous carotid artery commonly develop following a head trauma, after skull base surgery or in association with infections of the cavernous sinus. These lesions can rupture into the sphenoid sinus with occurrence of delayed epistaxis, and are often associated with a poor prognosis [4]. We describe a patient with a non-traumatic aneurysm of the intracavernous carotid artery that ruptured in the sphenoid sinus: this condition has seldom been reported in the literature. Following coiling of the lesion we observed an almost immediate recurrence of the lesion, in the form of a pseudoaneurysm occupying the sphenoid sinus.

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Figure 2 Control angiography after the embolization of the pseudoaneurysm with coils (A). Angiographic demonstration of the recurrence of the aneurysm, 20 days after embolization. Coil compaction inside the sac is evident (B). Obliteration of the lesion is achieved with a new coiling session (C).

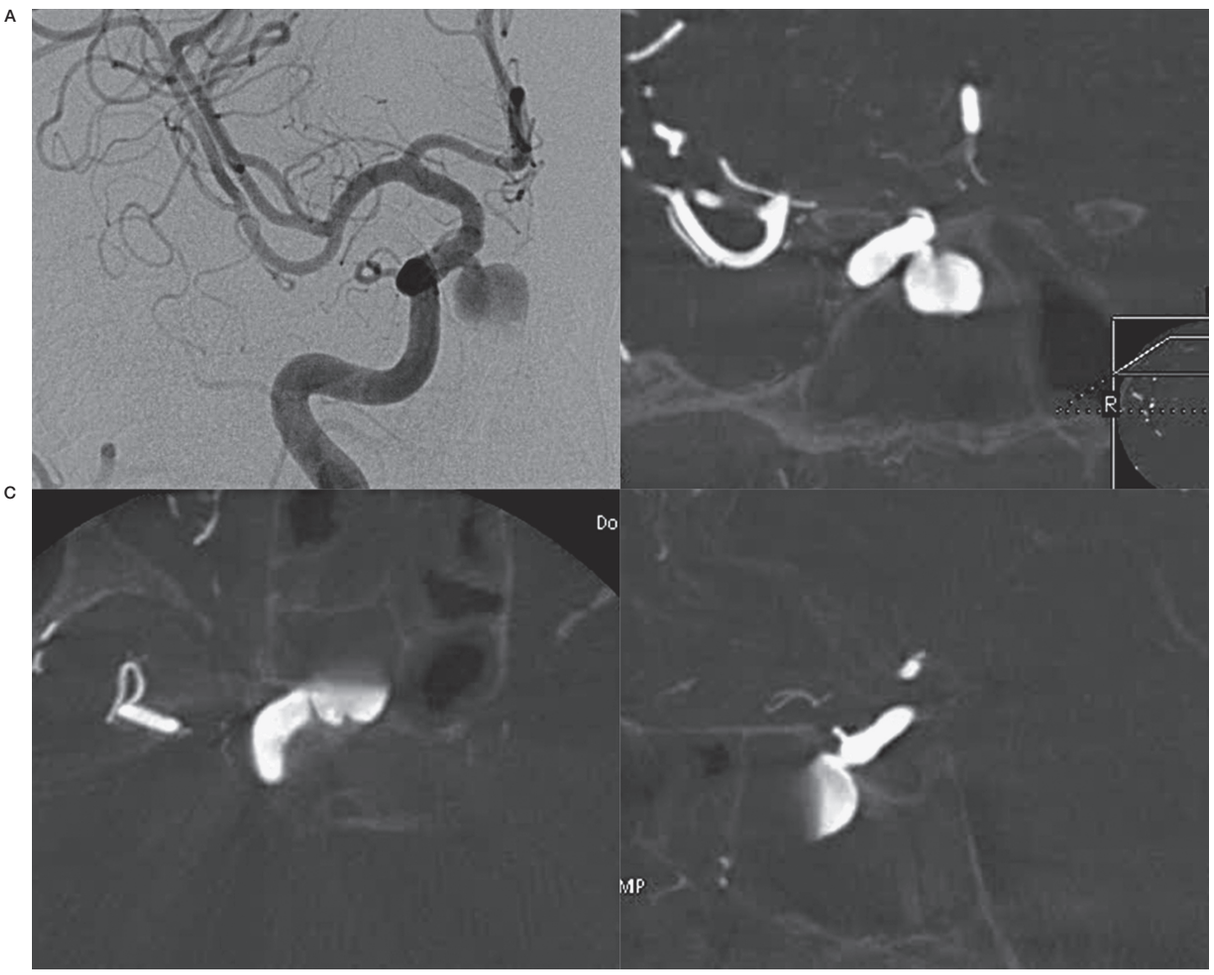
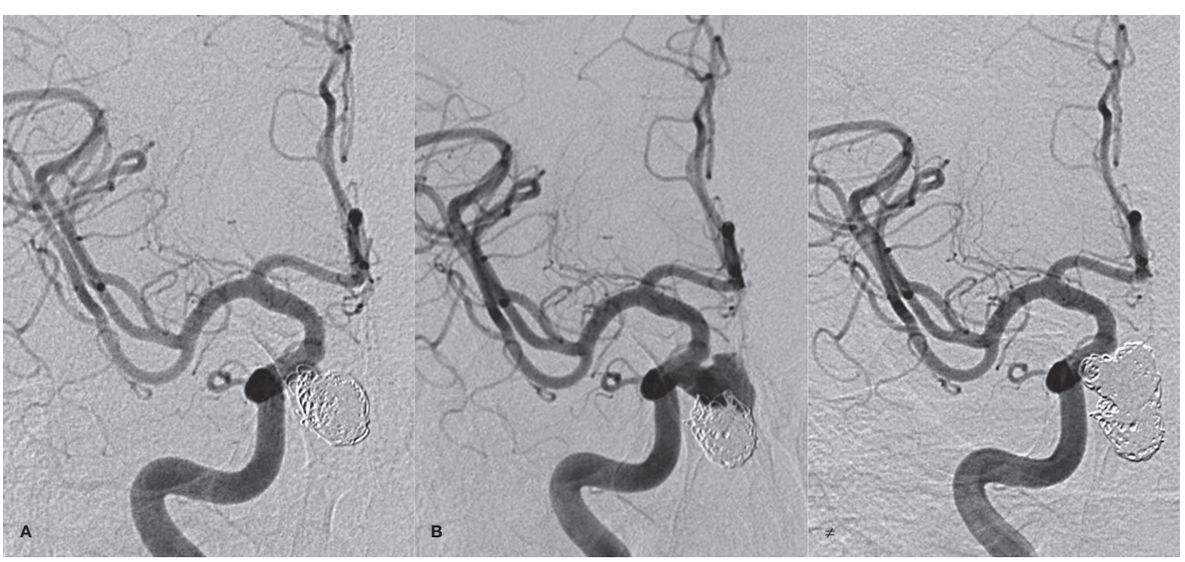


Figure 1 Conventional digital subtraction angiography on admission shows a pseudoaneurysm of the cavernous portion of the right internal carotid artery, with a small neck and "hazy" margins (indicative of endoluminal contrast stagnation and mural thrombosis). Multiplanar reconstructions after rotational acquisitions clearly depict the erosion of the lateral wall of the sphenoid sinus, which is completely occupied by a large clot.



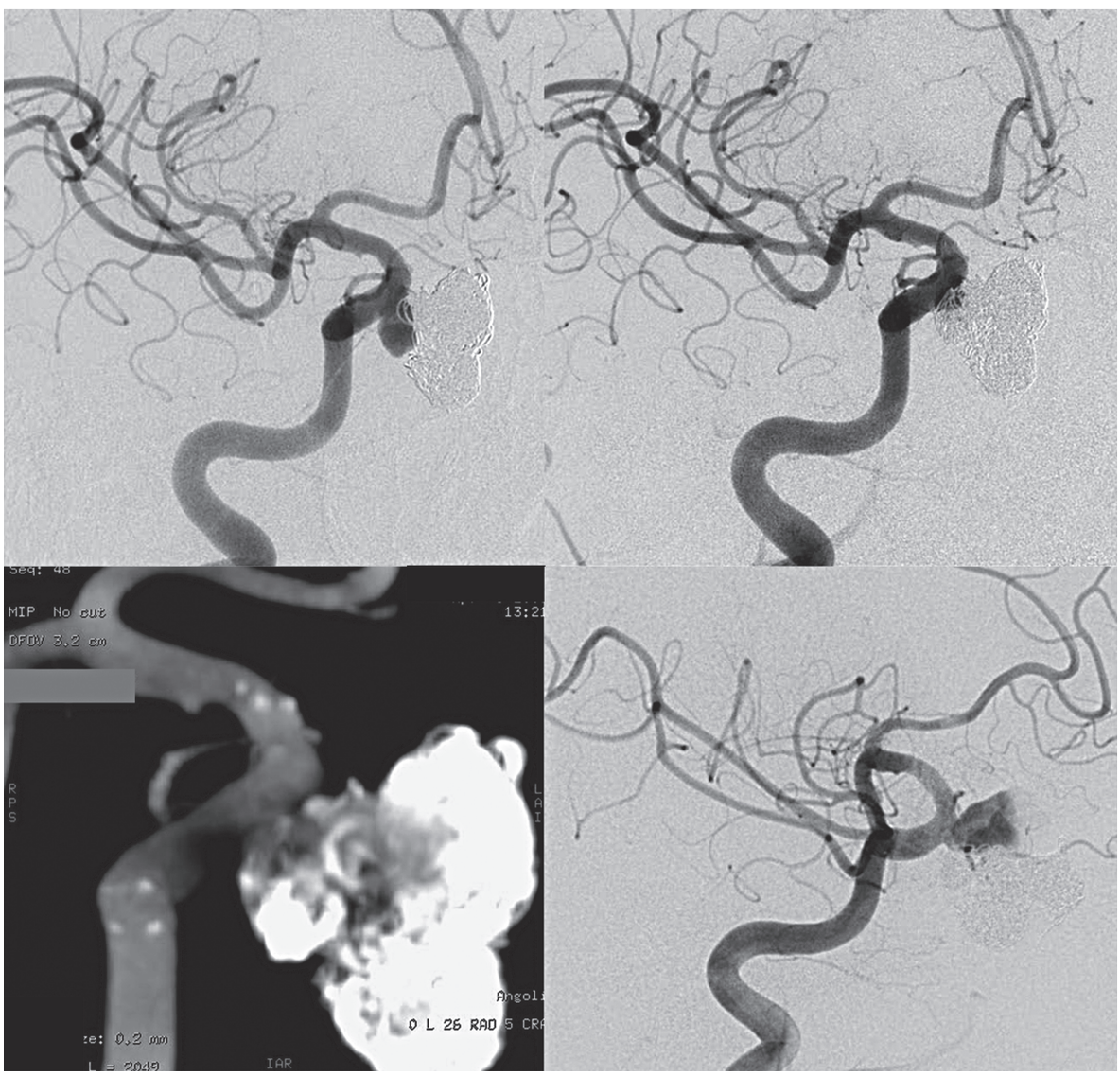


Figure 3 New recurrence of the lesion, 18 days after the second embolization (A). After treatment with coiling and stenting, a small residual opacification is evident at the aneurysmal neck (B). 3D reconstructions at the end of the procedure allow a good visualization of the coil packing and of the markers of the stent (C). Further recurrence is evident 25 days after the stenting, again associated with coil dislocation (D).

Case Report

Our patient was a 74-year-old man who had been suffering from recurrent right-sided epistaxis for nine days. No head trauma or skull base surgery were reported.

On admission to a nearby hospital blood chemistry revealed a hemoglobin value of 9 g/dl; nasoendoscopy revealed a throbbing clot in the right sphenoid sinus. Head CT scan and CT angiography confirmed the suspicion of an intracavernous carotid artery aneurysm with intrasphenoid extension and rupture. He was then transferred to our institution. Conventional angiography (performed in emergency) showed gravitational stagnation of contrast, "hazy" margins, and extramural thrombosis, suggesting a diagnosis of an intrasphenoid pseudoaneurysm (Figure 1). Endovascular coiling of the lesion was performed, and at the end of the procedure it appeared completely obliterated (Figure 2A).

Twenty days later the patient was admitted again for recurrence of nosebleed and angiography showed a major reopening of the pseudoaneurysm (Figure 2B): a balloon occlusion test of the right internal carotid artery showed only a partial cross compensation with a slight delay of the ipsilateral venous filling, so a new session of coiling was performed and the lesion was once again obliterated (Figure 2C). Nevertheless, fifteen days following this procedure epistaxis recurred, and MR angiography displayed a regrowth of the sac at the neck. While epistaxis was controlled with nasal packing, the patient was preloaded with aspirin and clopidogrel. Three days later, we performed angiography (Figure 3A) and attempted to place a covered coronaric stent across the neck of the sac, without success. So the new sac was coiled again and an intracranial self-expandable stent was placed in the neck. Post-procedural controls showed only a small residual opacification of the sac (Figure 3B,C).

After a new nosebleeding episode (twenty-five days following stent placement), MR angiography showed a large regrowth of the lesion, confirmed at conventional angiography (Figure 3D). At this time parent artery occlusion appeared the only therapeutic choice feasible in emergency to cure the patient: the lesion and the right internal carotid artery were then occluded with coils. Post-procedural angiographic controls showed a good cross-compensation of the blood flow through both the anterior and the posterior communicating arteries. Post-procedural MR displayed several small ischemic lesions in the right cortex and centrum semiovale. Epistaxis episodes stopped and our patient was dismissed one week later with a moderate left-sided paretic deficit. No aneurysmal reopening was observed after one year at MR angiography follow-up- Clinical follow-up shows a partial regression of the initial deficit, with a residual slight paretic deficit in the left upper limb.

Discussion

Epistaxis in an uncommon feature of nontraumatic intracavernous carotid artery aneurysms, and was described in one patient out of 37 by Linskey et al. [2]. Cases of epistaxis due to nontraumatic intracavernous carotid artery aneurysms with extension in the sphenoid sinus were recently reviewed by Lehmann et al. [5], who identified 36 cases reported in the literature. Data emerging from these reviews confirm that this condition is associated with a high mortality risk (about 22%) and that requires immediate therapy. Otherwise, in untreated cases the outcome is fatal in up to 71% of cases.

In our patient epistaxis was associated with the formation of a large partially thrombosed pseudoaneurysm occupying the right sphenoid sinus. Our initial approach was aimed at treatment of the lesion with preservation of the internal carotid artery, with coiling of the pseudoaneurysm first, and then using a stent-assisted technique. Both these strategies have proved their efficacy in several series and cases reported in literature [3,6]. Nevertheless, the recurrence of the bleeding and the reopening of the aneurysm in our patient made the sacrifice of the parent vessel unavoidable. A similar management has already been reported in patients harbouring a pseudoaneurysm of the carotid siphon with rapid growth and short-term recurrence after obliteration with coiling [7]. As a general rule, the factors most frequently involved in early aneurysmal recurrence after endovascular coiling are coil compaction and a product of neck diameter and inflow angle [8]. Undoubtedly these factors played a role in our patient, since some coil compaction was observed after the second coiling session, and the aneurysm had a wide neck and pointed forward, being prone to the water-hammer effect of the inflow bloodstream. In a series described by Sluzewski and Van Roij, the strongest risk factor for early rebleeding of coiled aneurysms was the presence of an adyacent hematoma [9]. They hypothesized that the thrombus located at the rupture site can evolve into a pseudoaneurysm and account for a high risk of recurrence and rebleeding. It is likely that in our patient such a condition was established in the closed environment of the sphenoid sinus: this may account for the disposition of the newly formed sacs at the periphery of the coil mesh, and without apparent relation with the neck of the aneurysm. As a consequence, the recurrent lesions had morphologies different from each other.

Struffert et al. [10] recently described two cases of post-surgical pseudoaneurysms of the cavernous internal carotid artery with intra-sphenoid extension, treated with coil embolization and preservation of the parent vessel. In both cases a delayed coil migration and dislocation occurred, suggesting that the embolization of these lesions may not be stable over time. The

authors claimed this may be related to the absence of the usual vessel wall elements in pseudoaneurysms: consequently, coils may exert their radial force against a compliant surrounding, and may expand or dislocate themselves into the sinusal cavity. Our report further supports these assumptions, and implies that early recurrence may also occur in this setting.

Several issues in our report deserve further discussion. First, a dual antiplatelet treatment in a patient with a ruptured aneurysm is risky and may be questionable. However, stenting of ruptured intracranial aneurysms has been widely described and the risks linked to this procedure are generally considered low and acceptable [11]. Second, since internal carotid occlusion was necessary in this patient, a question may be raised whether a bypass graft should have been performed in conjunction with the endovascular therapy. On the other hand, the real effect of this treatment is still not known conclusively [12].

In conclusion, our case report confirms that while epistaxis is a common presentation for traumatic aneurysms of the internal carotid artery, it can also be associated with intrasphenoid bleeding of an idiopathic aneurysm. Our description also supports the hypothesis that an intrasphenoid rupture of an intracavernous carotid artery aneurysm poses a high risk for the formation of a pseudoaneurysm due to the constrained environment in which the blood clots. Strict follow-up is warranted after treatment, since both early and delayed recurrence are possible. However, this condition may be untreatable with conservative endovascular treatment and may require the sacrifice of the ipsilateral internal carotid artery.

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